ELSEVIER

SPEECH-MOTOR AND LINGUISTIC SKILLS OF YOUNG STUTTERERS PRIOR TO ONSET

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Theorists have increasingly suggested that both speech-motor and linguistic factors are involved in the etiology of stuttering. This contention has been supported by findings that tend to indicate that youngsters who stutter have a slower speech rate and are less linguistically skilled than nonstutterers. However, no inferences can be drawn from these findings as to the nature or the causation of this disorder. This is because the aforementioned findings might be a result rather than a cause of the disorder. In order to clarify the directionality issue, a multi-year prospective study was undertaken that involved 93 preschool children with a parental history of stuttering.

At the initial session, none of the high-risk children sampled was regarded as having a stuttering problem. One year later, 26 children were classified as stutterers. Statistical analyses revealed that prior to the onset of stuttering these children did not differ from the other youngsters studied with respect to either their receptive or expressive language abilities. However, their rate of articulation was significantly faster. The latter finding is taken to mean that the children who developed stuttering were not limited in speech-motor ability. Rather, their fluency failures are seen as a result of a relatively high articulation rate. It is noteworthy, in this regard, that the rate of the high-risk children who continued to be viewed as nonstutterers was slower than that previously reported for youngsters of their age. This suggests that the slower rate served as a buffer against fluency breakdown.

INTRODUCTION

Central to a number of recent theories of stuttering is the assumption that both speech-motor and linguistic factors are involved in the etiology of the disorder (St. Louis, 1979; Van Riper, 1982). Various speech-motor and lan-

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guage studies have been carried out to test the assumed relationship between these factors and stuttering. With respect to speech-motor behavior, the evidence suggests that stuttering is associated with deficits in the individual motor subsystems involved in speech as well as in the coordination between these subsystems (Peters and Starkweather, 1990). Still more, comparative studies have shown that the speech-associated motor responses of adult stutterers are generally slower and more variable than those of nonstutterers (Adams, 1984; Janssen and Wieneke, 1987). Reaction-time studies also suggest that the lag is found both in the preparation or programming and in the physiological execution of their motor system for movement (Peters, Hulstijn, and Starkweather, 1989).

Though the speech-motor studies of adult stutterers have most often shown them to respond more slowly than nonstutterers do, this has not been the case with children; here the results have been equivocal. In some studies the results have been significant, while in others they have not. Though Meyers and Freeman (1985) observed that preschoolers who stutter have a significantly slower speech rate than those who do not, Richardson (1985), Kelly and Conture (1992), and Ryan (1992) found no difference in their speaking rate. Moreover, these contrasting findings do not stand alone. The results that relate to other acoustic measures, such as voice onset time (VOT), vowel duration, voice initiation (VIT), and voice termination time (VTT), are also inconclusive. Though Adams (1987), for example, observed that stuttering children have longer segment durations and slower voice onset times than nonstuttering youngsters do, other researchers have not found a significant difference in VOT and segment durations (Zebrowski, Conture, and Cudahy, 1985). Similarly, while some researchers have reported significant between-group differences in the VIT and VOT of stuttering and nonstuttering children (Cross and Luper, 1979, 1983), others have not (Murphy and Baumgartner, 1981; Winkler and Ramig, 1986).

The equivocal nature of the data that have resulted from comparative investigations of children who do and do not stutter are not limited to those that concern speech-motor behaviors. The studies that have been undertaken to investigate the role of linguistic factors in stuttering have also produced inconsistent findings. In this regard, the results of some studies suggest that children who stutter are generally less skilled linguistically than normally fluent children are. This is because they usually develop language skills more slowly than nonstutterers do and tend to use simpler linguistic structures than their fluent peers do (Kline and Starkweather, 1979; Wall, 1980). However, Ratner and Sih (1987) did not find that the children who stutter differed significantly in overall language performance from their nonstuttering peers.

The data from other studies have also made it apparent that care needs to be taken in commenting on between-group differences in the overall linguistic skills of stuttering and nonstuttering children. This is because of results of investigations like those of Byrd and Cooper (1989). They found that though children between the ages 5 and 9 who stutter were delayed in expressive language, they did not differ in receptive language from age-matched nonstutterers. This finding was supported by the observation by Ryan (1992) that most of the significant differences between preschool stuttering and nonstuttering children related to the expressive aspect of language.

The reported tendency of young stutterers to be delayed in their expressive language skills is generally interpreted as being a result rather than a cause of their disordered fluency. It is seen as a reflection of the stutterers' attempt to simplify their verbal responses as a means of coping with either anticipated dysfluency or the presence of fluency failure (Byrd and Cooper, 1989; Starkweather, 1987). This interpretation has flourished despite disparate results, like those of Westby (1979), who failed to find a significant difference in the expressive vocabulary of stuttering and nonstuttering children.

Though the aforementioned post-onset cross-sectional studies may be useful in generating hypotheses, it is questionable as to whether or not they are of much use in gaining knowledge about factors involved in the etiology of stuttering. Even if speech-motor and language studies consistently showed stutterers to differ from nonstutterers, the question would remain open as to whether the differences are the cause or result of the disorder. In order to address this issue and to test the assumptions that have been generated about the role of these variables, the results of longitudinal studies are needed. Unfortunately, data are lacking in regard to the speech-motor and linguistic functioning of children prior to the onset of stuttering. This is so for children of both sexes even though it would not be at all surprising to find gender differences in relation to these variables (Ryan, 1992; Walker, Archibald, Cherniak, and Fish, 1992).

It was the absence of the aforementioned longitudinal data that led to the present study. Because of the void, a prospective study was undertaken in which both male and female children with a family history of stuttering were studied for several years. The long-term objectives of the longitudinal project were to gain knowledge about the relation between genetic, physiological, linguistic, and environmental factors relating to the onset and development of stuttering among children whose fluency is at risk. The children studied were considered to be at risk because investigations have shown that approximately 20% of youngsters with a family history of stuttering are likely to become stutterers (Kidd, 1977; Kidd, Heimbuch, and Records, 1981; Kidd, Kidd, and Records, 1978; Van Praag and Janssen, 1980). In all likelihood, also, a majority of these stutterers would be male children (Kidd et al., 1978; Yairi and Ambrose, 1992).

The current report presents data from the first year of a three-year study of the speech-motor and linguistic skills of young children, considered to be at risk relative to fluency because of a family history of stuttering. One of the objectives of this investigation was to assess the speech-motor and linguistic skills of all of the sampled children in order to compare the skills of those who later develop into stutterers, with those who continued to be viewed as nonstutterers. Another objective was to explore further the relationship between gender, speech-motor and linguistic skills, and the onset of stuttering.

METHOD

Subjects

The subjects of this study were 93 monolingual, Dutch-speaking, preschool children. Each of them came from a different family in which one or both of the parents was a diagnosed stutterer. The stuttering parents whose children participated in this study were located through outpatient referrals from pediatricians and speech-language pathologists.

When the study began, the 93 children (45 boys and 48 girls) who made up the subjects sample were between the ages of 23 and 58 months (M = 39 months, SD = 9). At that time none of them was viewed as a stutterer by their parents or displayed signs of stuttering.

Sixty-five of the children in the study had a stuttering father, 23 a stuttering mother, and 5 had both a stuttering father and a stuttering mother. Both the children's pediatrician and the parents reported all of them were of normal intelligence and had no history of communication disorders. In addition, each of the participating children passed a pure tone audiometric hearing screening.

Procedures

Before both the initial test session and the follow-up, 1 year later, the parents were asked to report on their child's fluency by filling out the Disfluency Questionnaire (Kloth, Janssen, and Kraaimaat, 1989). This 10-item questionnaire measured on a 5-point Likert type scale that ranged from "never" to "very often" the extent to which normal (5 items), borderline (2 items), and stuttering types of speech disruption (3 items) were displayed at home during the previous 2 months.

Data collection for the initial test session and the year end follow-up of the children took place at the Department of Phoniatrics of the University Hospital of Utrecht. Two rooms separated by a one-way screen mirror were used. The subjects' room contained a selection of age-appropriate toys (e.g., two telephones, a bucket with small toys; Fisher-Price farmhouse, tea set), a Sony TC-158SD audiotape recorder, and a Grundig LC-290H video camera. In the observation room, there was a Philips VHS video recorder (type HQ-VR-722), a JVC color video monitor (type TM-210PS-K), and a remote control for the video camera.

At the start of the two test sessions, each of which lasted approximately 2 hr, a speech pathologist assessed the language development of the children by means of the Dutch versions of the Reynell Language Development Scale (Reynell, 1983; Bomers and Mugge, 1989), and the Peabody Picture Vocabulary Test (PPVT) (Dunn, 1965; Manschot and Bonnema, 1978). Then, each child's spontaneous speech was video recorded during 30 min of free-play with the mother. During this play period, the mother was instructed to converse and play with her child as she would at home.

Minutes 3 through 13 of each child's taped conversation with the mother during the initial and follow-up test sessions were transcribed by a trained coder. The 10-min conversations were separated into utterances using the Golinkoff and Ames (1979) criteria. That is to say, an utterance was defined as a string of words that communicated an idea, that was bound together by one breath, and that was intended to be continuous. Utterances were separated by pauses longer than 1 s.

The transcripts and the videotapes also were used to note the specific types of speech disruption that each child exhibited during the 10 min of conversation with his or her mother. The fluency failures observed were categorized as follows: normal disfluencies, defined as revisions, interjections, nontense silent pauses, phrase repetitions, or slow whole word repetitions; borderline disfluencies, defined as slow, nontense sound or syllable repetitions; and stuttering disfluencies, that is rapid sound or syllable repetitions, and tense silent or oral prolongations. Percentages of normal disfluent, borderline disfluent, and stuttering-like disfluent words were calculated by dividing each of these forms by the total number of words spoken by the child.

Data Analysis

Language Skills. For each subject, age-equivalent scores for receptive language development were obtained from the Reynell and the PPVT test procedures and for expressive language development from the Reynell. These scores were transformed into standard score equivalents. In addition, each child's mean length of utterance (MLU) during the transcribed 10-min spontaneous speech periods was used as a measure of his or her expressive language skill. MLU was determined by dividing the total number of nonrepeated words spoken by the child by the number of utterances. One-word utterances, stereotypical (e.g., thank you), or noninteractive phrases (counting and singing) were not a part of the data analysis (Brown, 1973).

Speech-Motor Skills. In order to assess the children's speech-motor skills, an acoustical temporal analysis was performed on the first 10 perceptually fluent utterances made by each of them during spontaneous speech that was equal to their individual mean length of utterance. MLU utterances that had poor audibility were not selected. The utterances were recorded at a sampling rate of 10 kHz and displayed as a time waveform with a duration of 2 s. Each utterance was measured from the onset of the periodic waveform of the first vowel or voiced consonant to the offset of the periodic waveform of the last vowel or voiced consonant. The simultaneous playback of the audio signal facilitated the identification of the onset and offset of the periodic waveform. Within an utterance, pause duration was defined as the absence of spectral energy between two words that exceeded 250 ms. Duration of each utterance in ms was automatically calculated following the positions of the cursors on the waveform. Also determined were the number of syllables within the measured utterances.

Articulation rate was calculated by dividing the duration of the utterances by the number of syllables that they contained. These values were then converted to syllable rate per second. The mean and standard deviation of the utterance durations across the 10 studied were computed for each subject. From these analyses, the children's mean articulation rate was derived. Articulation rate was defined as the duration in syllables per second of an utterance exclusive of pauses. Intrasubject variability in this measure was determined from the coefficient of variation, which was derived from the standard deviation divided by its mean (Kent and Forner, 1980).

Reliability

Intrajudge and interjudge reliability for identifying and classifying the type of fluency failures were calculated by the first and second author from a random selection of 10% of the speech samples using the Sander agreement index (Sander, 1961). Intrajudge reliability for identifying and classifying fluency failures was 93%. Interjudge reliability was 92%.

Interjudge reliability for the articulation rate measures were calculated from a random sample of 5% of the rate measures. Thus, 50 utterances were remeasured by a trained independent examiner. A Pearson's product moment correlation applied to these measures of rate revealed a coefficient of +.92.

RESULTS

At the time of the initial session, none of the 93 children was reported by the parents to display stuttering disfluencies and no such fluency failures were observed by the experimenters. However, at the follow-up session 1 year later, 26 children met the preset criteria that classified them as stutterers. Specifically, their parents had to indicate on the Disfluency Questionnaire (Kloth et al., 1989) that stuttering types of disfluency were often or very often displayed at home and both parents had to regard their child as a stutterer. For 16 of these 26 children, stuttering forms of disfluency also were observed in the child's speech during the follow-up session. The remaining 67 children were classified by their parents as normally fluent speakers who did not have a stuttering problem. Stuttering disfluencies were not evidenced by any of these children during the 1-year follow-up session.

The gender and the pre-onset mean age at initial testing of the 26 who, on follow-up 1 year later, were considered to be stutterers (i.e., experimental subjects) and the 67 who were not (i.e., control subjects), are shown in Table 1.

There it can be seen that 16 of the experimental subjects were male and 10 were female. The mean age of the males was 3 years 5 months and that of the females was 3 years 1 month. The control group consisted of 29 males with a mean age of 3 years 6 months and 38 females with a mean age of 3 years 2 months. The two groups did not differ to a statistically significant extent with respect to either age ($t = .34$, $p = .74$) or gender (Chi-square $= 2.5$, $p = .11$).

Table 2 shows the age-adjusted means and the standard deviations by gender for the initial speech-motor and linguistic measures of the experimental and control subjects. As can be seen, the former subjects showed, descriptively, a somewhat faster articulatory rate than the control group but much the same speech-motor variability.

To test for statistically significant differences between the subject groups, the sexes, and for group by gender interaction, each dependent measure was analyzed by means of a two-factor analysis of variance. Because the sample sizes were unequal, a regression procedure was used (SPSS, Tabachnick and Fidell, 1989). And, because age was found to correlate signifi-

Table 1. Gender and Pre-onset Age of the Experimental and Control Subjects

	Experimental subjects			Control subjects		
		Age (years; months)			Age (years; months)	
Gender	*n*	*M*	*SD*	*n*	*M*	*SD*
Male	16	3;5	;11	29	3;6	;8
Female	10	3;1	;9	38	3;2	;11
Total	26	3;3	;10	67	3;4	;10

Table 2. Age Adjusted Means and Standard Deviations for Initial Speech-Motor and Linguistic Measures of the Experimental (n = 26) and Control Subjects (n = 67)

	Experimental subjects				Control subjects			
	Male		Female		Male		Female	
	M	*SD*	*M*	*SD*	*M*	*SD*	*M*	*SD*
Speech-motor								
Articulation rate	3.70	.61	3.84	.41	3.46	.56	3.53	.56
Variability art. rate	.23	.08	.20	.05	.20	.05	.23	.09
Language receptive								
Reynell	.12	1.04	.06	.89	−.01	.90	.30	.72
PPTV#	108.35	13.38	102.12	15.51	109.39	16.70	106.24	15.49
Language expressive								
Reynell	−.24	.91	−.49	.91	−.22	1.02	.08	.69
MLU	4.19	.99	4.04	.77	4.66	1.02	4.40	1.25

PPVT-scores were available for only 18 of the experimental and 53 of the control subjects.

cantly with the speech-motor and MLU measures, it was used as a covariate in the analyses of these variables. The results of the analyses are shown in Table 3.

With respect to the articulatory rate across gender, the between-group F values shown in Table 3 indicate that at the time of initial contact the articulation rate of the children who a year later were categorized as stutterers differed significantly from that of the control children. That is to

Table 3. Two-Factor Analyses of (Co)variance for Groups, Gender and Group by Gender Interactions

	Two-way ANCOVA F-values			
	Covariate age	Group	Gender	Group × Gender
Speech-motor				
Articulation rate	50.71**	8.28**	3.06	.22
Variability art. rate	16.26**	.02	.01	3.56
Language receptive				
Reynell	–	.30	.87	.87
PPTV	–	.24	1.02	.30
Language expressive				
Reynell	–	1.69	.33	1.94
MLU	61.16**	3.56	1.74	.09

$^*p < .05$; $^{**}p < .01$

say that prior to showing signs of stuttering they had significantly faster articulation rates than the children who on follow-up continued to be seen as nonstutterers.

As the descriptive data relative to the variability of articulation rate suggested, the groups did not differ to a statistically significant extent. Similarly, the group-by-gender interaction failed to reach statistical significance. This is consistent with the fact that the boys, who a year later were viewed as incipient stutterers, were only slightly more variable in their articulation rate during the initial test session than the girls who, on follow-up, would be similarly categorized. It is noteworthy, however, that the opposite was the case for the nonstuttering control group.

With regard to pre-onset linguistic skills, the between-group F values found in Table 3 make it apparent that the receptive language ability of the children who on follow-up were considered to be stutterers did not differ significantly from that of the control subjects on either the Reynell or the PPTV. The groups also failed to show a significant difference with respect to either the Reynell or the MLU measures of expressive language.

DISCUSSION

Ninety-three children with a parental history of stuttering were the subjects of this prospective study. Though, initially all were viewed as nonstutterers, follow-up a year later revealed that 26 were then regarded as stutterers. This finding is consistent with the genetically based high-risk hypothesis that has been proposed over the years in relation to children of stutterers (Andrew and Harris, 1964; Van Praag and Janssen, 1980; Kidd, 1977; Kidd et al., 1981). Of those children who came to be seen as stutterers, 16 were male and 10 were female. This yielded a male-to-female ratio of 1.6:1, which is larger than that found by Yairi (1983) in a younger group of stuttering children (1.2:1) but smaller than the estimated 4:1 ratio among adult stutterers (Bloodstein, 1987).

The main question of this study was whether or not the speech-motor and language skills of children whose fluency is at risk are related to the development of stuttering. The strongest result of this prospective study is that the pre-onset articulatory rate of the 26 children who on follow-up a year later were considered to be stutterers differed from that of the 67 who continued to be viewed as nonstutterers. Specifically, their pre-onset articulation rate was significantly faster than that of the control children. The two groups did not differ in the variability of their articulation rate, however.

In contrast, research with adults who stutter has shown them to be consistently slower and more variable in the initiation and execution of speech movements (Adams, 1984; Janssen and Wieneke, 1987; Peters et al., 1989). A relatively high variability has been thought to be indicative of a less well-developed speech-motor system (Kent and Forner, 1980; Cherniak and Schneiderman, 1986). This, together with the slower speaking rate among adult stutterers, has led researchers to theorize that stuttering is the result of organismic limitation. However, the current finding of a higher pre-onset articulatory rate among those who were later seen as stutterers and the absence of a between-group difference in variability is inconsistent with this hypothesis. Rather, the finding of a relatively high pre-onset articulation rate among the experimental subjects is more in line with Bloodstein's (1987) notion that high speaking rates can result in stuttering and with Starkweather's (1987) suggestion that stuttering among children may result from a rate that exceeds their motor-control capacity. To the extent that this is the case, at least for the group of genetically vulnerable children studied, a mean articulatory rate of 3.75 syll/s would seem to have been too fast for them to maintain normally fluent speech.

Of interest with respect to the aforementioned hypotheses is whether or not the mean pre-onset articulation rate of the high-risk children who were later seen to be stutterers deviates from that of a reference group of children who do not stutter. In this regard, Walker et al. (1992) found the mean articulation rate of normally fluent 3- and 5-year-old children to be 3.82 syll/s and 4.28 syll/s, respectively. For a group of nonstuttering boys, who ranged in age from 4 to 5 years 10 months, Meyers and Freeman (1985) reported a mean articulation rate of 4.04 syll/s. These normative data for English-speaking children are comparable to those of Dutch youngsters (Haselager, Sliss, and Rietveld, 1991). The mean articulation rate of a group of 5-year-old children was found to be 4.03 syll/s. These normative data would seem to suggest that it is the capacity and not the articulatory rate of the experimental subjects that is deviant. For these children, that is, even a relatively slow articulatory rate apparently exceeded the threshold needed for the maintenance of fluency. Consistent with this is the fact that the pre-onset mean articulation rate of the high-risk group of children that did not show signs of stuttering on follow-up was 3.45 syll/s. This rate is notably slower than that of the 3-year-old, normally fluent children in the Walker et al. (1992) study. Possibly, then, it was the relatively slow speech of our 3-year-old nonstutterers that served as a protective factor against fluency breakdown.

The present study was also directed at the pre-onset receptive and expressive language skills of both stutterers and nonstutterers. The results suggest that before onset the children who later were considered to be stutterers were not different from the at-risk children who remained nonstutterers with respect to either their receptive or expressive language skills. This would seem to indicate that language deficiency was not an etiologic factor for those high-risk children who were seen as stutterers by the time they reached approximately 4 years of age. This does not, however, preclude the possibility that language difficulties may be of influence in later onset or that it might not affect the persistence of stuttering. Follow-up evaluations of the high-risk children who were the subjects of this multiyear longitudinal investigation will shed some light on these issues.

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